

Cervical Osteophytosis As A Rare Cause of Dysphagia: A Case Report

Nadir Bir Disfaji Nedeni Olan Servikal Osteofitozis: Olgu Sunumu

DENİZ BELEN, LEVENT GÜRSES

Social Security Hospital, Department of Neurosurgery, Ankara

Received : 4.11.2000 ⇔ Accepted : 10.1.2001

Abstract: Cervical osteophytes are frequently found in geriatric patients. Although it is uncommon to observe dysphagia induced by these osteophytes, this is an important and treatable condition. The diagnosis may be established using plain radiographs of the spine, esophagography, computed tomography, or magnetic resonance imaging. We describe the case of a patient with severe dysphagia who had a large anterior cervical osteophyte. The differential diagnosis, treatment options, and pathophysiology of the process are discussed.

Key words: Diffuse idiopathic skeletal hyperostosis (DISH), dysphagia, Forestier's disease, spinal osteophytosis

Özet: Servikal osteofitler ileri yaşta sıkça karşılaşılan bir durumdur. Servikal osteofit sonucu ortaya çıkan disfaji oldukça nadir olmasına rağmen tedavi edilebilir bir durumdur. Tanı; omurga kolonunun düz grafisi, özefagogram, BT veya MRG ile konulabilir. Bu yazıda servikal osteofite bağlı şiddetli disfajisi olan bir hasta sunularak ayırıcı tanı, tedavi yöntemleri ve patofizyoloji tartışıldı.

Anahtar kelimeler: Diffüz idiopatik iskelet hiperostozisi (DISH), disfaji, Forestier hastalığı, spinal osteofitozis

INTRODUCTION

Although dysphagia and cervical spondylosis are both common presenting problems, they are often unrelated. A review of the literature showed that dysphagia caused by this type of bony change is relatively rare, with fewer than 200 cases reported to date (4,11). The association of these two problems was first reported in 1926 by Mosher (4). In 1950, Forestier described a clinical manifestation that was characterized by spinal rigidity, and in the 1970s this was named „diffuse idiopathic skeletal hyperostosis“ (DISH). Most reported cases of spinal

osteophyte-related dysphagia have been in DISH patients (4,10).

The diagnostic criterias for DISH, also known as Forestier's disease, are as follows: 1) flowing calcification/ossification along the anterolateral aspect of four contiguous vertebral bodies 2) relative preservation of intervertebral disc height in affected areas and 3) absence of apophyseal joint ankylosis and sacroiliac joint sclerosis/fusion (10). The etiology is unknown, but there is no relationship between this condition and ankylosing spondylitis, degenerative disc/joint disease, or rheumatoid arthritis. The

majority of cases are asymptomatic. The overall prevalence of DISH is higher than would be expected in the predominantly white population over 50 years of age, and the frequency is lower in the African-American, Native-American, and Asian populations, suggesting that there may be a genetic origin (13). A report claimed that Middle Eastern populations exhibit less dysphagia symptomatology than United States and European populations, which have higher frequencies of DISH (1). It is uncommon to diagnose this disease in patients less than 50 years of age. The prevalence increases with age, to a maximum of 28% in individuals 70 to 80 years of age. Also, there is a gender bias towards men in those affected, with a male:female ratio of 2:1 (4).

CASE DESCRIPTION

A 68-year-old man presented with the complaint of painful swallowing of 2 years' duration. The problem had gradually worsened over the previous 6 months, and he had recently developed problems swallowing even liquid food. The patient

had lost approximately 10 kg of body weight during the past 6 months. His laboratory findings were unremarkable, apart from a slightly elevated blood-glucose level of 150 mg/dL. Gastrointestinal specialists attempted esophagoscopy assessment, but this was unsuccessful due to a high-level obstruction of the esophagus. A plain x-ray of the cervical region showed extensive calcification along the anterior aspect of the cervical column from the C2 to C5 vertebrae (Figure 1). A barium-swallow esophagogram revealed mechanical obstruction caused by this same lesion, but showed no other pathology (Figure 2). Cervical magnetic resonance imaging (MRI) revealed details of the lesion and the displacement of the surrounding soft tissue (Figure 3). There were no significant degenerative osteophytic changes within the spinal canal or the foraminae of the cervical vertebrae, nor was any calcification of the posterior longitudinal ligament. Plain radiographic screening of other parts of the skeletal system revealed no additional osteophytic degeneration.



Figure 1: A lateral plain film of the cervical spine shows a large anterior cervical osteophyte extending from C2 to C5. The trachea is displaced ventrally.



Figure 2: A lateral-view barium-swallow esophagogram demonstrates a high-level esophageal obstruction.



Figure 3: Sagittal cervical MRI reveals the same pathology that was detected on plain radiographs, but without pathologic enhancement. There are no obvious osteophytic changes in the spinal canal or vertebral foraminae.

Since the patient was having severe problem of swallowing even liquid food, and in light of the progression of the disease, we advised that the compressing mass should be surgically removed. However, he refused surgical treatment and was placed on a special liquid diet. Six months later, his weight had stabilized and he was able to swallow the prescribed recipe.

DISCUSSION

DISH is a common skeletal condition that can affect the entire spinal column, but the two most frequently affected regions are the thoracic and cervical spine, respectively (10). Cervical spondylosis is recognized as a rare cause of dysphagia, aspiration pneumonia, myelopathy, cervical root compression, upper airway obstruction, dysphonia, and vertebral artery compression. Symptoms are particularly prevalent in patients with DISH, who may have large cervical osteophytes. Aspiration is common in

individuals with dysphagia who have cervical osteophytes larger than 10 mm diameter (11).

Regarding other regions, thoracic spondylosis has been cited as a cause of myelopathy, Horner's syndrome, radiculopathy, thoracic outlet syndrome, obstructive pneumonia, and esophageal food impaction. Similar to the above situation, symptoms are more likely to arise in patients with DISH, who have large bridging osteophytes. Lumbar osteophytes in DISH patients have been implicated in lumbar spinal stenosis and obstruction of the inferior vena cava. DISH is also associated with diabetes mellitus, with extraosseous calcification in the tendons and peripheral ligaments, and with heel, shoulder, and elbow pain. Bony fractures are also a feature in this patient group (6).

The diagnosis of DISH is based on radiological assessment of the thoracic spine, although extraspinal involvement can also help identify the disease (7). Plain x-rays are the first step in patient evaluation. The spinal radiological manifestations of DISH are characterized by extensive ossification of the anterolateral aspect of the vertebral bodies, with relative preservation of disc height. Bridging ossification is most common in the thoracic spine, and non-bridging ossification is characteristic of cervical and lumbar involvement. Ossification of the posterior longitudinal, intraspinous, nuchal, and flavum ligaments has been reported as well (7). The next diagnostic step may be MRI or CT, which allow the examiner to investigate the pathology in detail and plan the surgical approach. A barium-swallow esophagogram is indicated in cases of cervical involvement to assess esophageal function.

When a patient known to have a cervical vertebral bone spur complains of dysphagia, it must be determined whether the protuberance is the singular cause of the dysphagia, a contributing factor, or whether it plays any role at all. Alternative causes of dysphagia must be considered, including neurological diseases (stroke, Parkinson's disease, and amyotrophic lateral sclerosis) and/or mechanical obstruction due to head/neck cancer, mediastinal masses, Zenker's diverticulum, esophageal webs, and stricture or cancer of the esophagus (8). In addition, gastroesophageal reflux can cause symptoms similar to those seen with cervical bony protuberances.

Asymptomatic patients with cervical vertebral bony changes or cases where a problem is detected incidentally may be approached conservatively. The

natural course of the disease is slow, and surgery is associated with significant morbidity and mortality. Sometimes it is difficult to intubate these individuals due to their rigid cervical spine, and fiberscope guidance can help in these cases. Some clinicians prefer to intubate while the patient is awake (9). The most widely accepted treatment for cases of severe dysphagia is surgical removal of the compressing mass, but the problem does not always resolve immediately after the operation. Some authors have claimed that the dysphagia is caused by a combination of mechanical pressure, chronic esophageal wall inflammation, and dysfunction of the nerve supply to the wall due to stretching. McCafferty et al. (8) reported that their patients' dysphagia had continued long after surgical treatment, whereas others have found that the problem resolved immediately (3).

Another issue of debate regarding surgical management of these cases is the need for spinal stabilization after the mass removed. Some authors have pointed out that the process is slow, the spine is rigid (10), and elderly people are at greater risk of developing problems during or after any long surgery. These investigators believe that cervical fusion does not help these individuals, and that the restricted neck motion after fusion may cause them to suffer even more (8). In contrast, most published data have shown that cervical fusion after osteophyte removal is essential for stabilization (2,3,4). Our opinion is that the decision to perform any fusion procedure should be made on a case-by-case basis.

Today, various disciplines are attempting to manage this pathology with significantly different approaches. Some orthopedic surgeons and ear-nose-throat specialists do not hesitate to treat cervical osteophytic lesions without neurosurgical consultation (5,12), but their surgical methods are not necessarily what a neurosurgeon would advise. Neurosurgeons frequently deal with problems in the cervical spine, and are very experienced with the pathologies that affect all structures in this region. In our opinion, patients with cervical vertebral osteophytes who are surgical candidates should always be consulted by a neurosurgeon.

Correspondence: Deniz Belen,
SSK Ankara Eğitim Hastanesi,
II. Nöroşirürji Kliniği, Dışkapı 06150,
Ankara, Turkey
e-mail: denizbelen@hotmail.com

REFERENCES

1. El-Garf A, Khater R: Diffuse idiopathic skeletal hyperostosis (DISH). A clinical study of the disease pattern in Middle Eastern populations. *J Rheumatol* 11(6): 804-807, 1984.
2. Epstein NE: Simultaneous cervical diffuse idiopathic skeletal hyperostosis and ossification of the posterior longitudinal ligament resulting in dysphagia or myelopathy in two geriatric North Americans. *Surg Neurol* 53: 427-431, 2000
3. Goffin J, Calenbergh FV: Forestier's Disease. *J Neurosurg* 85: 524-525, 1996.
4. Granville LJ, Musson N, Altman R, Silverman M: Anterior cervical osteophytes as a cause of pharyngeal stage dysphagia. *J Am Geriatr Soc* 46(8): 1003-1007, 1998
5. Grasshoff H, Motsch C, Mahfeld K: Vertebragenic dysphagia. *Zentralbl Chir* 124: 1041-1044, 1999
6. Le Hir PX, Sautet A, Le Gars L, Zeitoun F, Tubiana JM, Arrive L, Laredo JD: Hyperextension vertebral body fractures in diffuse idiopathic skeletal hyperostosis: a cause of intravertebral fluidlike collections on MR imaging. *AJR* 173: 1679-1683, 1999
7. Mata S, Chhem K, Foertin PR, Joseph L, Esdaile JM: Comprehensive radiographic evaluation of diffuse idiopathic skeletal hyperostosis: development and interrater reliability of a scoring system. *Semin Arthritis Rheum* 28: 88-96, 1998
8. McCafferty RR, Harrison MJ, Tamas LB, Larkins MV: Ossification of the anterior longitudinal ligament and Forestier's disease: an analysis of seven cases. *J Neurosurg* 83: 13-17, 1995
9. Palmer JH, Ball DR: Awake intubation with the intubating laryngeal mask in a patient with diffuse idiopathic skeletal hyperostosis. *Anaesthesia* 55(1): 70-74, 2000.
10. Rotes-Querol J: Clinical manifestations of diffuse idiopathic skeletal hyperostosis (DISH). *British J Rheumatol* 35: 1193-1196, 1996
11. Strasser G, Schima W, Schober E, Pokieser P, Kaider A, Denk DM: Cervical osteophytes impinging on the pharynx: importance of size and concurrent disorders for development of aspiration. *AJR* 174(2): 449-453, 2000
12. Uppal S, Wheatley AH: Transpharyngeal approach for the treatment of dysphagia due to Forestier's disease. *J Laryngol Otol* 113(4): 366-368, 1999.
13. Weinfeld RM, Olson PN, Maki DD, Griffiths HJ: The prevalence of diffuse idiopathic skeletal hyperostosis (DISH) in two large American Midwest metropolitan hospital populations. *Skeletal Radiol* 26: 222-225, 1997